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A Case Report of Acute Obstructive Suppurative Cholangitis in a Non-O-1 *Vibrio Cholerae* Biliary Carrier

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Summary

A case report is presented of a 73-year-old male who was seen with fever, jaundice, abdominal pain and central nervous system depression. He failed to respond to intensive antibiotic therapy, and subsequently acute obstructive suppurative cholangitis fully developed. Upon laparotomy, the patient's gallbladder was found to be enlarged with the bile from the gallbladder and bile duct itself containing a high pus content. Its cultured organism revealed non-O-1 *Vibrio cholerae*. To our knowledge, no prior case of acute obstructive suppurative cholangitis in a non-O-1 *Vibrio Cholerae* biliary carrier has been reported in Japan.

Introduction

Acute obstructive suppurative cholangitis (AOSC) is relatively rare, but has a very high mortality rate. In the present paper, we report on our emergent operation on an AOSC patient, in whom non-O-1 *Vibrio cholerae* and *Bacteroides fragilis* are cultured from the intraoperatively aspirated bile of the gallbladder and choledochus. These non-O-1 *Vibrio cholerae* and *Bacteroides fragilis* had a good possibility of the cause of the AOSC. The paper discusses the first non-O-1 *Vibrio cholerae* culturing from the bile duct in Japan.

Case Report

A 73-year-old male was admitted to our institution due to fever, jaundice, and right upper quadrant pain. He had been well until four years previously, when he underwent an operation for duodenal bleeding. Since the age of 72, he had been receiving psychological treatment for night delirium and disorientation. Two months prior to being admitted to our institution, the patient first experienced fever (38°C), vomiting, and urinary incontinence. However, his continued severe night delirium resulted in temperature developed, and his consciousness became drowsy. Through emergency ultrasound examination of the abdomen, he was diagnosed as having acute cholangitis, and was admitted to our care.

Key words: Acute obstructive suppurative cholangitis, Non-O-1 *Vibrio cholerae*, Bile duct, Jaundice, Monotricha.

索引語: 急性閉塞性化膿性胆管炎, non-O-1 *Vibrio Cholerae*, 総胆管, 黄疸, 単毛菌.

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Table 1. Laboratory Data on admission.

WBC	11900/uL	Cholinesterase	0.68
RBC	333/uL	Alk. phosphatase	49 U
Hemoglobin	10.4g/dl	Cholesterol	146mg/dl
Hematocrit	31.0%	GOT	73 U
		GPT	123 U
Serum Protein	6.0g/dl	LDH	253 U
Albumin	2.8g/dl	rGTP	190 U
Globulin	3.2g/dl	LAP	99 U
Total Bilirubin	4.1mg/dl	Sodium	138mEq/l
Direct Bilirubin	2.6mg/dl	Potassium	3.8mEq/l

On admission, his consciousness was drowsy. On examination of the abdomen, tenderness and muscle tightening in the right upper quadrant were present. As seen in Table 1, leucocytosis and liver dysfunction were detected. An enlarged gallbladder with a thickened wall was recognized on ultrasound examination. Although no calculus was present, there was slight debris in the gallbladder. The choledochus was 10mm in diameter and the intrahepatic bile duct was 3mm in diameter. Figure 1 shows that both of these are not dilated. Since acute obstructive suppurative cholangitis could not be denied, we performed an urgent laparotomy under general anesthesia. We found the gallbladder to be enlarged, with the bile from the gallbladder being pus itself, white in color and tinged with green. Bile from the choledochus was revealed to be the same. Both were aspirated using an aseptic procedure (Fig. 2). After cholecystectomy, a cholangiography was performed. We found complete obstruction of the distal bile duct, the so-called reversed U form. (Fig. 3). On palpation, there was the tumor which its consistency was elastic hard with a size of thumb finger head. Considering the

**Fig. 1** Ultrasound Examination.

Arrow: debris in the enlarged gallbladder.

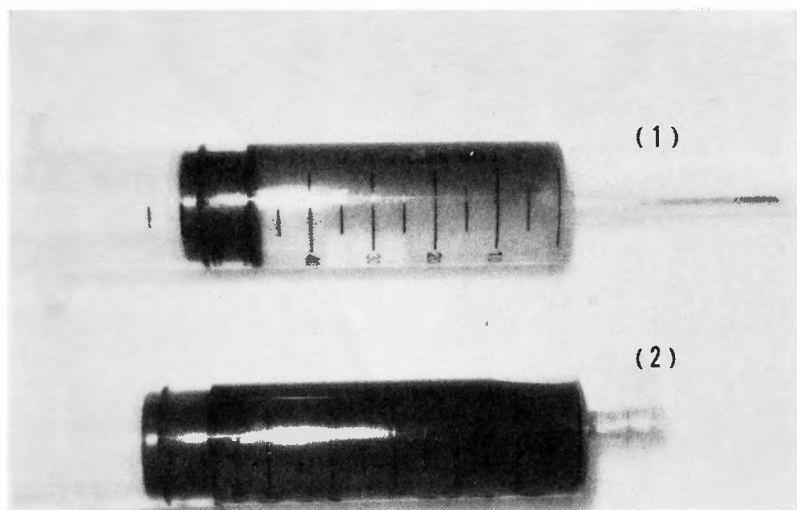


Fig. 2 Bile of the gallbladder (1)
Bile of the choledochus (2)



Fig. 3 Operative cholangiography.

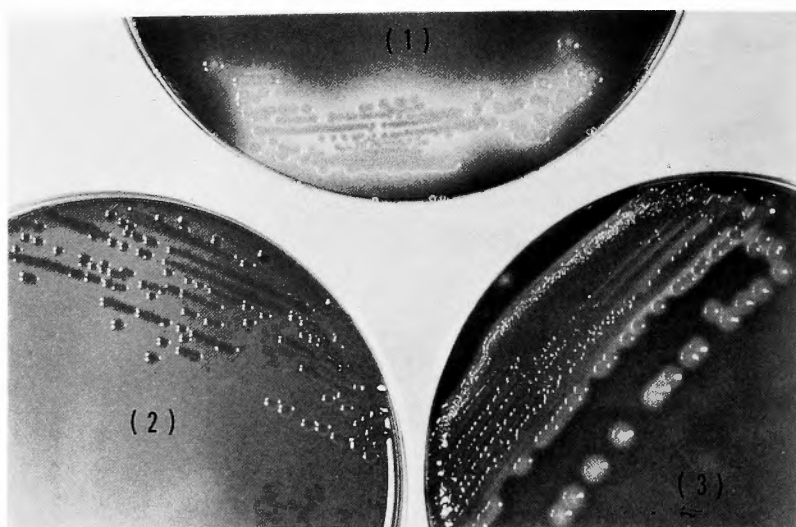


Fig. 4 1) Modified Drigalski agar.
2) TCBS culture medium.
3) Sheep hemolytic culture medium.

emergency operation and postoperative problems such as delirium at night, however, we decided to perform a choledochoduodenostomy (side to side) and tube drainage of the distal bile duct. After surgery, Latamoxef, Tobramycin, Clindamycin and Gentamicin were used as antibiotics for 9 days. The organisms cultured from the bile of the drainage tube became negative. The clinical findings and laboratory data returned quickly to normal.

The organisms cultured from the intraoperatively aspirated bile of both gallbladder and choledochus were determined to be non 0-1 *Vibrio Cholerae* and *Bacteroides fragilis*. A brief

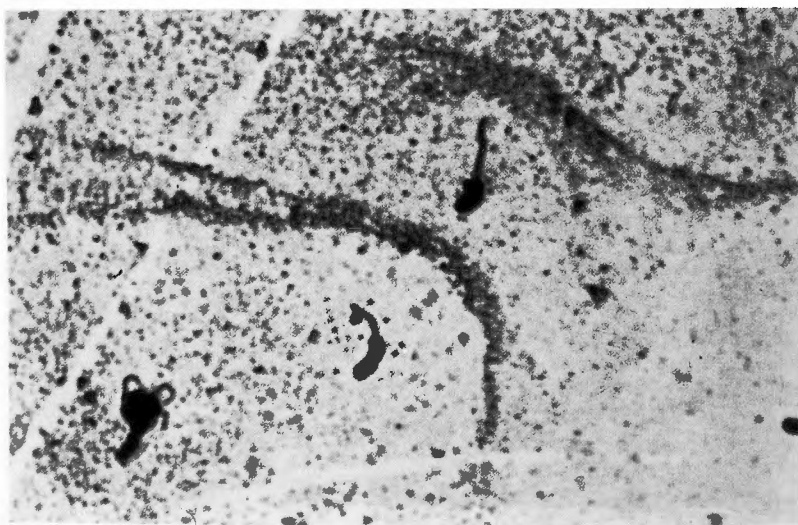


Fig. 5 Monotricha which is detected using the flagellar stain (HE stain).

explanation of this determination process follows. First, we confirmed the halophilic and hemolytic bacteria by Modified Drigalski agar and sheep hemolytic culture medium. Since the colony was formed in the TCBS culture medium (Fig. 4) and Monotricha was detected in a flagellar stain (Fig. 5), we then confirmed the culture to be *Vibrio cholerae*. We subsequently studied the biological disposition. Glucose dissolution discounted *Minicus cholerae*. Finally, the lack of coagulation in the 0-1 anti-serum confirmed our suspicion that the culture was non-0-1 *Vibrio cholerae*. In terms of pathological findings, a considerable number of neutrophils was found to have widely and deeply invased the wall of the gallbladder, with moderate fibrosis found in its submucosa. Incidentally, non-0-1 *Vibrio cholerae* organisms were not isolated from a patient's stool.

During the three months of postoperation, endoscopic retrograde cholangiography (ERCP) evidenced normal papilla of Vater and that there was communication between the narrow distal bile duct and duodenum. The patient were discharged from our hospital on the two postoperative weeks. And he is doing on the eleven postoperative months.

Discussion

The criteria responsible for generating acute cholangitis has been differently reported thus far. The generally accepted criteria of AOSC involves 5 signs, however: the so-called CHARCOT'S triad¹⁾, fever, abdominal pain and jaundice plus shock and central nerve system damage as presented by REYNOLDS⁵⁾ in 1959. Our case presented all signs but shock.

SHIROTA et al.⁸⁾ reported that the mortality rate of AOSC was 82.9% by conservative therapy, and 44.5% by biliary tract drainage. We thus consider drainage of the biliary tract as soon as possible to be very important. In this way, we confirmed this patient got well.

According to the statistical study in our country, the total number of organisms cultured from the bile among the 4075 cases of cholelithiasis confirms that aerobic bacteria is found in 88% and anaerobic in 12%. (Table 2). Among the highest detected rate of aerobic bacteria is *E. coli* at 40.7% and *Klebsiella* at 33.1%. Among anaerobic organisms, *Bacteroides* constitute 21.7%⁹⁾. Many of these complex patterns exist, especially, for example, among the elderly, the aerobic and anaerobic bacteria complex represents 50%³⁾. As can be seen, an average of 3.9 kinds of organisms were detected⁴⁾. Among the 139 cases of AOSC, cultured organisms from the bile duct were *E. coli* (74.8%) *Klebsiella* (61.9%) *Pseudomonas* (25.2%) *Enterobacter* (18.7%), and *Strept. faecalis* (10.1%). It is important to note that non-0-1

Table 2. Organisms cultured from the gallbladder of cholelithiasis (Shirota A⁸⁾.

Cultured Organism	Bile of the gallbladder	Bile of the choledochus
aerobic bacteria only	1819 (92.7%)	656 (91.2%)
aerobic bacteria plus anaerobic bacteria	115 (5.9%)	62 (8.6%)
anaerobic bacteria only	28 (1.4%)	1 (0.2%)
Total	1962	719

Vibrio cholerae was not detected. It is characteristics that in our case, non-O-1 *Vibrio cholerae* was cultured from the intraoperatively aspirated bile. We confirm that non-O-1 *Vibrio cholerae* and *Bacteroides fragilis* were the important factors that caused AOSC.

Non-O-1 *Vibrio cholerae* is different from O-1 *Vibrio Cholerae*, that is, genuine cholerae, in terms of somatic antigens. Originally, non-O-1 *Vibrio cholerae* was said to thrive in the tropics, and was not found in Japan until 1960. Now, however, non-O-1 *Vibrio cholerae* strains widely distributed in the environment in Europe, Asia, and the United States. Studies of the ecology of non-O-1 *Vibrio cholerae* have demonstrated that these organisms are found in brackish surface waters, are more numerous during warmer months⁶⁾. But, Enterotoxin, which is a strong pathogenicity, is rare produced by non-O-1 *Vibrio cholerae*. According to SAKAZAKI⁷⁾, the production rate of enterotoxin is said to be 16%. JAMES M. HUGHES²⁾ reported that non-O-1 *Vibrio cholerae* infection is not a reportable disease in the United States, and is probably often not diagnosed when it does occur, non-O-1 *Vibrio cholerae* infection undoubtedly represents a small portion of the actual total. Infections outside the gastrointestinal caused non-O-1 *Vibrio cholerae* are rare, with only 18 cases having been reported throughout the world. Among the 18 cases, organisms were cultured from the bile, gallbladder, blood, wound, ear, sputum, appendix vermiformis, ascites and CSF⁷⁾.

This is perhaps the first time that biliary infection caused by non-O-1 *Vibrio cholerae* has been seen in Japan. Furthermore, this case exhibited stenosis of distal bile duct without stone. Following surgery, this stenosis improved. Importantly, however, the relationship between the temporal obstruction of the bile duct and the infection of non-O-1 *Vibrio cholerae* remains uncertain.

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和文抄録

起炎菌が NON-O-1 *Vibrio Cholerae* であった
急性閉塞性化膿性胆管炎の一例

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症例は73歳の男性で発熱と黄疸と意識レベルの低下を来たして受診した。抗生剤治療に反応せず、急性閉塞性化膿性胆管炎は進行していった。緊急で開腹した。胆嚢は緊満腫大し、胆嚢内および総胆管内の胆汁は青白色の膿汁で満ちていて、その培養で non-o-1 *Vibrio*

Cholerae が検出された。術後第一病日で菌は陰性となり、経過は良好であった。おそらく本胆管炎症例は、胆管内から non-O-1 *Vibrio Cholerae* が検出された本邦最初の手術治験例であろう。